A Rare Case of Vallecular Cyst

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ABSTRACT
Vallecular cyst is an uncommon but potentially dangerous condition causing stridor and has been associated with sudden airway obstruction resulting in death due to its anatomical location in neonates. It may also present with feeding problems resulting in failure to thrive. Endoscopic laryngoscopy is necessary to visualize vallecular cyst when suspected clinically. Other conditions leading to neonatal stridor should be ruled out, such as laryngomalacia and other laryngotracheal abnormalities. Marsupialization with a CO2 laser is the most successful treatment.

CASE REPORT
A 6-month-old male infant was referred to the ear, nose and throat (ENT) department of Dr. Susheela Tiwari Hospital, Government Medical College, Haldwani, for the evaluation of inspiratory stridor, feeding difficulty, suprasternal retraction, and failure to thrive. He was born at 38 weeks of gestation by vaginal delivery at a local hospital, weighing 2.10 kg, without perinatal problem. There was hoarseness when he cried, but he did not have significant respiratory distress. After 4 months he developed noisy breathing, and some days later stridor developed. The patient’s father consulted the pediatrician of a private hospital and treatment started along the lines of pneumonia and the patient was shifted to the pediatric intensive care unit. After 2 weeks of treatment, the patient was discharged. Few weeks later, the patient again developed stridor. He was brought to the emergency room and then admitted to the neonatal intensive care unit for treatment. After nasal continuous positive airway pressure (CPAP) support, severe respiratory distress was relieved. However, symptoms, such as hoarseness, noisy breathing, and feeding difficulty persisted.

He was evaluated with several studies, including chest computed tomography, but there was no abnormal finding present. Ten days after weaning from nasal CPAP, there was a sudden desaturation episode again, which was relieved by positive pressure ventilation via manual bag masking. For evaluation of upper airway, he was transferred to our department. On admission, his body weight was 2.10 kg, height was 49 cm, and head circumference was 36 cm. His body temperature was 37.1°C, pulse rate was 172/min, respiratory rate was 41/min, and blood pressure was 74/42 mm Hg. Conjunctivae and sclerae were within normal limits. The chest expansion was symmetrical, but there was inspiratory stridor and suprasternal retraction. Heart beats were regular without any audible murmur. The abdomen was soft and flat with normal bowel sound. Liver and spleen were not palpable.
There was no rash or petechiae on skin exam. All cranial nerve function were intact.

The results of laboratory test were hemoglobin 12 gm/dL, white blood cell count 7000/μl with a normal differential cell count, and platelet count 410,000/μl. The chest X-ray was normal. On rigid laryngoscopic examination, an anteriorly displacing cystic mass on the tongue base left side was detected. Magnetic resonance imaging (MRI) neck was done, which showed retention cyst about 1.6 × 1 × 1.2 cm in anteroposterior, transverse, and superoinferior dimensions in the left vallecula insinuating into the left base of the tongue, medially abutting the epiglottis (Fig. 1).

Under sedation, laryngoscopy confirmed the cystic mass between the base of the tongue and the epiglottis. Intubation was tried by an anesthesiologist, but the tube was not negotiable. Cyst was aspirated first and then intubation was done. After that, cyst was removed using bipolar cautery (Figs 2 and 3). After cautery, the position of the epiglottis was recovered. Removed specimen was sent for histopathological examination.

Immediately after surgery, inspiratory stridor, supra-sternal retraction, and feeding difficulty improved gradually. One week later, follow-up rigid laryngoscopy showed no abnormal finding except episodic influx of epiglottis. He was discharged and follow-up was done in the outpatient clinic. He was healthy at home, had no airway and feeding problems, and gained weight steadily.

**Histopathological Report**

Histopathologic examination showed a cyst lined by squamous epithelium with chronic active inflammation (hematoxylin and eosin, ×40) (Fig. 4).

**DISCUSSION**

Laryngeal cysts are a uncommon cause of stridor in infants. They are more commonly seen in adults. They have been classified based on location, histology, size, and contents. The vallecula is the depression behind the base of the tongue between the median and lateral epiglottic folds on each side. Vallecular cysts belong to
the category of ductal cysts described in DeSanto’s early classification system of laryngeal cysts.\(^2\) They classified laryngeal cysts into ductal, saccular, and thyroid foraminal subtypes. Ductal cysts are caused by mucus retention in the submucosal collecting ducts.\(^2\) Vallecular cysts may contribute to upper airway obstruction by posterior displacement of the supraglottis.

Clinical presentations of vallecular cysts include stridor, feeding problems, failure to thrive, gastroesophageal reflux, apnea/cyanosis, chest retractions, hoarse cry, as well as respiratory distress and life-threatening events.\(^3\) Laryngomalacia can coexist with vallecular cysts.\(^1\) Diagnosis is by direct laryngoscopy, though imaging studies, particularly MRI, can provide important information.\(^4\) Management is by transoral surgical removal or marsupialization.

About 60% of children with stridor have laryngeal obstruction, such as laryngomalacia, vocal cord paralysis, subglottic stenosis, hemangioma, or laryngeal cysts; 25% have lesions in the upper airway, including choanal atresia, macroglossia, facial anomalies; 15% are due to tracheobronchial lesions, such as tracheomalacia or vascular compression, and others.\(^5,6\) In 1881, Abercrombie provided the first description of a laryngeal cyst.\(^7\) Mitchell et al in 1987, published the largest single series consisting of 20 cases experiencing of the Hospital for Sick Children, London over a 15-year period.\(^5\) In 1980, Holinger reported on 219 children presenting with stridor to two Chicago pediatric units over four years. Two of these cases (0.9%) were due to laryngeal cysts.\(^9\) A total of 12 to 45% of laryngomalacia presents with other associated airway abnormality, such as laryngeal cyst.\(^5\) Thus, in neonatal stridor, evaluation of the airway anatomy and differential diagnosis from other causes of stridor are important to prevent any mortality and morbidity from this cause.

Newman classified laryngeal cysts as epithelial, tonsillar and oncocytic cysts (1984: Modified working classification).\(^10,11\) Two major hypotheses to explain the pathogenesis of vallecular cyst are that this cyst is a consequence of either ductal obstruction of mucus glands or an embryological malformation.\(^5\) Histologically, the cyst contains respiratory epithelium with mucus glands, with an external lining of squamous epithelium.\(^12\) Flexible laryngoscopic or bronchoscopic exam is usually performed to diagnose the vallecular cysts.\(^4\) Surgical removal may be the treatment of choice, alternative modalities, such as endoscopic marsupialization, excision and de-roofing have been developed recently.\(^1\) Simple aspiration of the cyst is not advised because of its high recurrence rate.\(^8\)

In our case, vallecular cyst was diagnosed in a 6-month-old male infant with symptoms of noisy respiration, recurrent upper respiratory tract infection (URT1). Patient was misdiagnosed and treated conservatively by pediatrician.

**CONCLUSION**

When an infant presents with noisy breathing or stridor, a complete ENT examination is required to rule out upper airway abnormalities. Awareness and a high level of suspicion are required for a quick diagnosis of such a lesion as it can predispose an infant to recurrent URT1, feeding problems, respiratory distress, or failure to thrive.

**REFERENCES**